CASE REPORTS

ISOLATED PULMONARY MUCORMYCOSIS IN AN APPARENTLY NORMAL HOST: A CASE REPORT

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Mucormycosis is a rare fungal disease commonly affecting individuals with diabetes mellitus, hematological malignancy, and immune deficiency. Isolated pulmonary mucormycosis is extremely rare. This article reports a case of isolated pulmonary mucormycosis that presented as a solitary cavity infiltrate in a patient with no underlying risk factors. (*J Natl Med Assoc.* 1995;87:572-574.)

Key words • pulmonary mucormycosis • cavitary infiltrate • amphotericin

Pulmonary mucormycosis is a rare and almost invariably fatal disease that usually occurs in the context of a hematological malignancy or a host immune deficiency state. It is extremely rare in normal individuals. Only 12 cases of mucormycosis have been reported in the normal host.[1-12] This article reports another case of an isolated pulmonary mucormycosis in an apparently normal host.

CASE REPORT

A 68-year-old black male was admitted with complaints of dyspnea and productive cough. The patient was apparently healthy until 1 day prior to admission. He was treated as an outpatient with theophylline and a B_2 agonist inhaler for bronchial asthma. There was no history of hemoptysis, fever and chills, night sweats, chest pain, diabetes mellitus, or weight loss. The patient denied using steroids.

Physical examination showed a well-developed afebrile black male in moderate respiratory distress. His blood pressure was 110/80 mm Hg, pulse rate was 92 beats/minute, and respiratory rate was 26 breaths/minute. Skin and oral mucous membranes were dry. Lung examination was remarkable for bilateral inspiratory wheeze. Heart and abdomen examinations were within normal limits. Lymph nodes were not palpable.

An arterial blood gas showed pH 7.49, pCO_2 29 mm Hg, and a pO_2 108 mm Hg on oxygen supplementation at 2 l/min. A white blood cell count was 13 500/mm^3, and hemoglobin, hematocrit, and platelet count were within normal ranges. Serum electrolytes and blood sugar values were within normal limits. Blood urea nitrogen and creatinine were elevated to 51 mg/dL and 2.4 mg/dL, respectively. A chest radiograph showed an oval-shaped cavitary infiltrate in the right upper lobe (Figure 1). A chest radiograph taken 1 month earlier had been normal.

Blood urea nitrogen and creatinine rapidly returned to normal with intravenous hydration. Dyspnea and wheezing resolved with routine treatment for bronchospasm; however, the lung infiltrate remained unchanged after 10 days of broad-spectrum antibiotic therapy. A sputum culture revealed light growth of *Candida albicans.* A computed tomography scan of the chest confirmed a localized right upper lobe density with cavitation and necrosis (Figure 2). The patient showed a positive reaction to intradermal injection of a purified protein derivative (PPD) tuberculin solution. However, three sputum specimens did not grow acid-fast bacillus after 6 weeks' incubation. A

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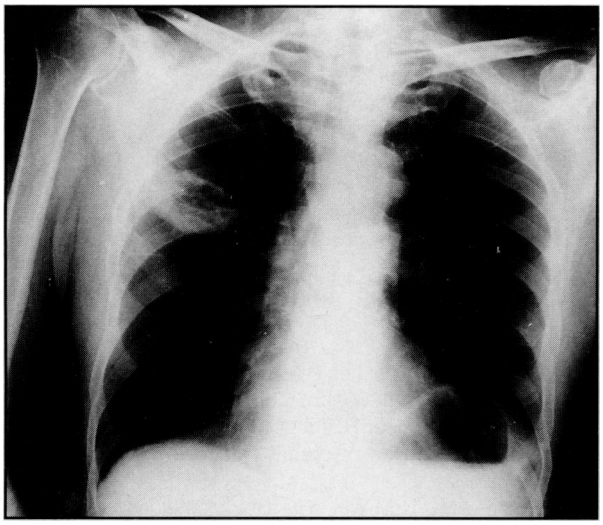

Figure 1. Chest radiograph showing right upper lobe cavitary infiltrate.

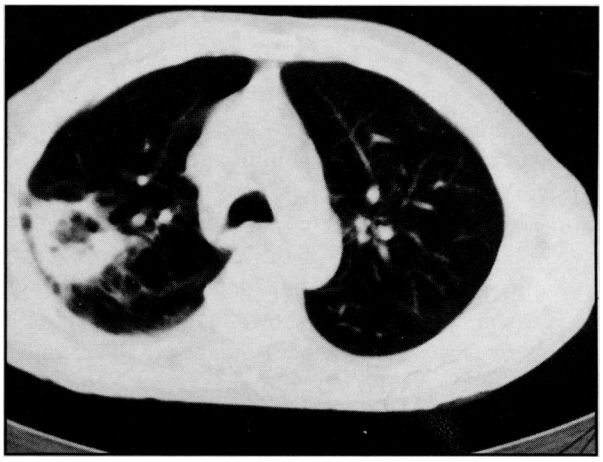

Figure 2. Computed tomography scan showing cavitary lung lesion.

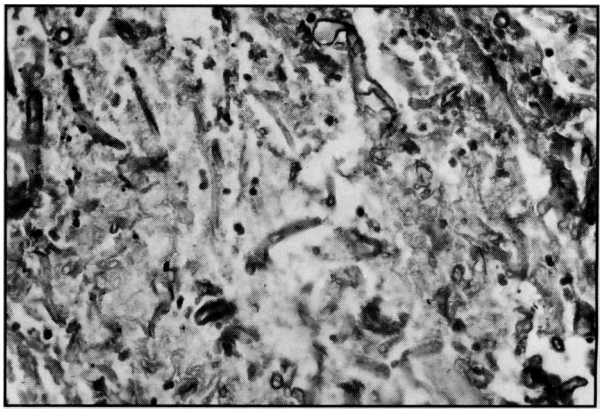

Figure 3. Lung biopsy specimen showing thick fungal hyphae of mucor.

transbronchial biopsy of the affected lesion showed thick fungal hyphae with right-angle branching and inflammation (Figure 3) highly suggestive of mucormycosis. A ratio of helper to suppressor T cells was 2.98, with an absolute helper T cell count of 2100 cells. The patient was started on amphotericin B, initially at 0.5 mg/kg per day, which was subsequently increased to 1 mg/kg per day. After several days of treatment with amphotericin B, the patient developed progressive renal insufficiency, which was attributed to the amphotericin B. The patient had received a total of 900 mg of amphotericin B before it was discontinued and fluconazole was started. Multiple blood cultures showed no growth. The patient was clinically stable until this time despite no improvement in the pulmonary infiltrate. The patient's general condition deteriorated thereafter, and he developed septic shock due to enterococcal sepsis that was complicated by adult respiratory distress syndrome, disseminated intravascular coagulation, and gangrene of both lower extremities. The patient subsequently died as a result of bacterial sepsis.

DISCUSSION

This is a unique case in that mucormycosis presented as a single cavitary lung infiltrate without symptoms related to the cause of the infiltrate in an immunocompetent host with no underlying risk factors. Mucormycosis refers to several different diseases caused by the fungi of the order Mucorales. The order Mucorales includes a family called Mucoraceae, which in turn, includes various genera such as *Mucor, Rhizopus, Rhizomucor,* and *Absidia*. More recently, zygomycosis is used in place of mucormycosis, which also includes other related fungi with similar morphology. The clinical manifestations and therapy for all these organisms are, however, identical.[13,14] These organisms are ubiquitous, saprophytic, and not fastidious. Their optimal temperature of growth is 28°C to 30°C under aerobic conditions, with an incubation period of 2 to 5 days.[13,14]

Incubation begins with inhalation of the spores or their direct inoculation into an abraded skin. This is followed by dissemination in patients with underlying diseases such as diabetes mellitus (especially with ketoacidosis), hematological malignancies, patients receiving chemotherapy or steroids, and in patients with underlying immune deficiency. Some rare causes of mucormycosis include chronic renal insufficiency[15,16]

and metabolic acidosis due to chronic salicylate poisoning.[17]

Clinical manifestations of mucormycosis can be divided into six different forms that include rhinocerebral, cutaneous, pulmonary, gastrointestinal, central nervous system, and a miscellaneous form involving bones, breasts, mediastinum, and kidneys. In order to cause the disease, spores must overcome the host's immune defense mechanisms. Experimental studies have shown that mice on steroid therapy or with streptozotocin-induced diabetes mellitus exhibit a specific defect in macrophage function allowing proliferation of the spores of Mucorales.[18-20] Common to all forms of mucormycosis is vascular invasion with tissue necrosis and infiltration of the tissue with neutrophils. Diagnosis is achieved by demonstrating broad nonseptate hyphae with right-angle branching in a tissue biopsy specimen. Rhinocerebral is the most common form of presentation for all varieties of mucormycosis. Isolated pulmonary mucormycosis is extremely rare and reported in the literature as case reports only.

Pulmonary mucormycosis has been shown to occur in various clinical forms.[21] It usually occurs as a solitary infiltrate rapidly progressing into life-threatening pneumonia. Rarer forms include endobronchial lesion and complications related to airway occlusion. Hemoptysis commonly occurs with vascular invasion. The right lung is more commonly involved than the left.

The treatment of pulmonary mucormycosis usually requires a combined medical and surgical approach. Review of the literature reveals little success with medical therapy alone. High-dose amphotericin along with surgical resection of the involved areas of the lung and treatment of the underlying disease is the mainstay of treatment. Kutsuzawa et al[12] reported therapeutic success using a combination of parenteral miconazole and inhaled amphotericin. Various other authors have reported successful usage of newer antifungal agents such as fluconazole and itraconazole.[11,22]

CONCLUSION

Despite various reports, high-dose amphotericin and early surgical intervention remain the therapy of choice for pulmonary mucormycosis at this time. The major determinant of the outcome is the status of the underlying disease.

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